Tungiasis: consequences of delayed presentation/diagnosis

Tungiasis is an endemic ectoparasitosis in the poor communities of South and Central America, sub-Saharan Africa, Asia, and the Caribbean countries.[1] The female jigger flea, *Tunga penetrans*, measuring approximately 1 mm in length, can penetrate any part of the body with a preference for the periungual region.[1,2] Tungiasis can occur in those wearing sandals and not just in those who walk in bare feet. Early diagnosis reduces the possibility of superimposed bacterial infection and complications including erysipelas, tetanus, nail loss, ulceration, fissures, chronic lymphedema, tissue necrosis, autoamputation, and gangrene of digits.[3–5] We report herein an unusual case of tungiasis resulting in serious clinical consequences.

A 45-year-old male farmer presented with a 5-month history of lesion on his third left toe. He reported a 'burning sensation' without history of insect bite, tenderness, and fever. A previous biopsy had reported an infected corn and the patient had been given quinolone antibiotic with symptomatic treatment. Presently he was using topical fusidic acid. On examination, there was a 1×1 cm^2 scabbed ulcer with surrounding edema and black discoloration at the base of the toe. Needle puncture caused purulent discharge to exude from the lesion relieving the pain. A tentative diagnosis of cutaneous leishmaniasis was made and parenteral Glucantime® was administered for a week. Differential diagnoses of malignancy, retained foreign body, and arthropod infection were considered.

Medication proved ineffective and the lesion grew increasingly painful. The third left toe was amputated and submitted for investigation. Histological examination revealed benign stratified squamous epithelium with keratosis. Body parts of an arthropod, prominently cuticle, were identified on cross section. Therefore a diagnosis of arthropod infection, tungiasis, was made (Figures 1 and 2).[6]

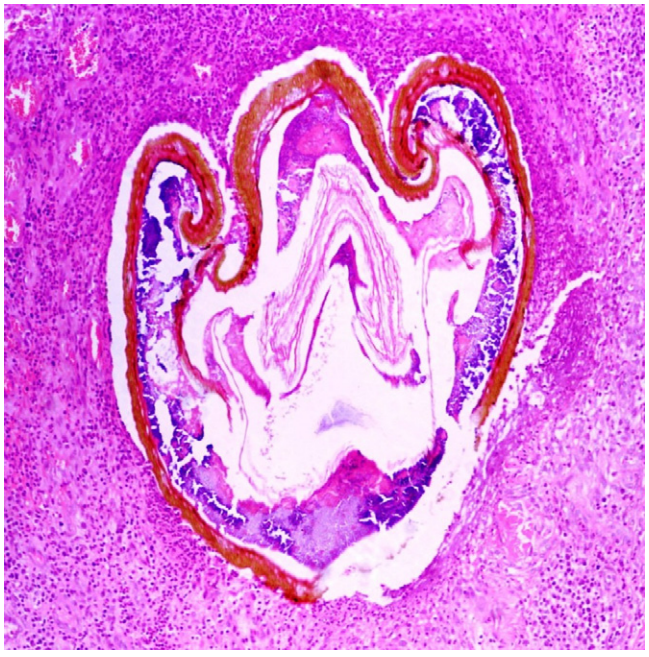

Figure 1 Cross section of the lesion showing Tunga flea with well-developed cuticle and other body parts surrounded by dense acute pyogenic inflammatory cell infiltrate.

Figure 2 A characteristic *Tunga penetrans* lesion (thick arrow), with pale-yellow papule and dark center, and a less obvious lesion (thin arrow) with surrounding chronic skin changes and multiple, loosely attached eggs.[6]Swaminathan A, et al. Tungiasis in recently arrived African refugees. *Med J Aust* 2005;**183**:51. © Copyright 2005. *The Medical Journal of Australia* — reproduced with permission.

Tungiasis can be misdiagnosed as warts, granulomas, tropical ulcers, scabies, tick bites, acute paronychia, ecthyma, secondary pyoderma, and abscesses, or even as malignant melanoma. Diagnostic incision procedures result in secondary cellulitis, erysipelas, tetanus, or septicemia.[7] While clinical presentation and diagnosis are typical in few cases, patients often present with lesions altered by external manipulation.[8] Early recognition and clinical diagnosis is therefore of prime importance in endemic areas to initiate prompt treatment and prevent complications. Clearly there were several errors in the diagnosis and management of this case, emphasizing the need for clinicians to have a high index of suspicion for tungiasis based on clinical appearance of the lesion. We conclude that tungiasis may mimic other serious conditions resulting in erroneous diagnosis and treatment.

Conflict of interest: No conflict of interest to declare.

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Maxillofacial Rosai—Dorfman disease in a newly diagnosed HIV-infected patient

Rosai—Dorfman disease is an uncommon benign proliferation of hematopoietic and fibrous tissue. Its initial manifestations most often include a roughly symmetric, painless, bilateral cervical adenopathy, fever, leukocytosis, and hypergammaglobulinemia, although extranodal disease may develop.[1—3] Rosai—Dorfman disease in HIV-infected patients has been previously reported in just three cases.[4—6] We report herein the case of a young Venezuelan woman, recently diagnosed with HIV, who had developed Rosai—Dorfman disease with maxillary and malar involvement.

A 56-year-old woman was evaluated at the ear, nose and throat (ENT) service with a three-year history of hard palate swelling with compromise of the right genian region. Physical examination revealed a volume increase in the right side of the face. A considerable hard palate swelling was observed. Laboratory studies on presentation revealed HIV infection (HIV-1 and -2 ELISA tests were positive, and infection was confirmed with the Western-blot test). Multiple lesion biopsies were undertaken. Other laboratory studies were performed; no alterations in complete blood count or chemistry were evidenced. Serology for *Coccidioides immitis*, hepatitis B virus, Epstein—Barr virus (EBV), and cytomegalovirus were positive. The CD4 cell count was 350 cells/μl and the viral load was 10 000 copies of RNA/μl. CT-scans showed a significant compromise of the soft tissues in the right maxillary region (including a significant compromise of bone structures). A subtotal therapeutic maxillectomy and biopsy were carried out. The maxillary antrum lateral wall was found to have fibrohistiocytic lesions and inflammatory changes. In the resected tissues, a significant number of large, pale histiocytic cells that contained apparently engulfed lymphocytes or plasmocytes within their cellular borders was observed (emperipolesis; Figure 1). These distinctive large, pale cells — Rosai—Dorfman cells — were S-100 protein-positive by immunostaining and so differ from ordinary histiocytes (Figure 1). CD68 immunohistochemistry was also positive.

Most cases of Rosai—Dorfman disease occur during the first or second decade of life, but any age group can be affected. The youngest patient on first series had congenital sinus histiocytosis with massive lymphadenopathy (SHML), and the oldest developed symptoms at age 74.[7,8] However, Rosai—Dorfman disease in HIV-infected patients has been previously reported in just three cases,[4—6] the first described in 1991. To our knowledge, this case is the fourth to be reported. Microscopically, there was a pronounced dilatation of the lymph nodes (see Figure 1). The sinuses were occupied by numerous histiocytic cells with a large vesicular nucleus and abundant clear cytoplasm, which may contain lipids and also lymphocytes and plasma cells. The histological key feature of Rosai—Dorfman disease is the presence of various numbers of large, pale histiocytic cells that contain within their cellular borders apparently engulfed lymphocytes or plasmocytes (emperipolesis); these distinctive large, pale cells — Rosai—Dorfman cells — are S-100 protein-positive by immunostaining and so differ from ordinary histiocytes.

Despite its sometimes impressive clinical presentation, Rosai—Dorfman disease is a benign and self-limiting disease, whose treatment is aimed largely at controlling local manifestations (most often by surgical therapy). The microscopic differential diagnosis, particularly in extranodal disease, is at times challenging and can include Langerhans cell histiocytosis, Hodgkin's disease, non-Hodgkin's lymphoma, metastatic carcinoma, and metastatic malignant melanoma. Rosai—Dorfman disease with maxillary compromise has been previously reported in four cases (non HIV-infected patients),[9—12] and no cases have been reported with malar compromise. In Venezuela, the first case of primary osseous Rosai—Dorfman disease was observed by us in 2002,[13] and to the best of our knowledge this pathology has not been reported again until our current case.